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CASE REPORT

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Alcohol injection for nonsurgical management of tailgut cyst in a middle-aged woman: A case report

Farrah Aljuhani | **Bashair Almunami** | **Rawan Alsamahi** | **Nadim Malibary** | **Zuhoor Algaithy**

Faculty of Medicine, King Abdulaziz University, Jeddah, Saudi Arabia

Correspondence
Nadim Malibary, Faculty of Medicine, King Abdulaziz University, abdullah sulayman, Jeddah 21441, Saudi Arabia.
Email: nadim.malibary@gmail.com

Abstract

Managing recurrent tailgut cyst in a patient who is refraining from the definitive surgical en bloc resection can be challenging. Therefore, in this case report we outlined a less invasive approach which is computed tomography-guided aspiration with alcohol injection which resulted in prolongation of symptoms free period in our patient.

KEYWORDS

alcohol injection, case report, tailgut cyst

1 | INTRODUCTION

Presacral tumors represent clinical challenges from diagnosis to management, mainly due to the rarity of these lesions, their nonspecific presentation, and the variety of their embryological origin. The presacral space is the space where fusion of the embryologic hindgut and neuroectoderm occurs. It contains totipotent cells from which various tumors may arise.[1] Presacral space tumors are mainly classified as congenital, neurogenic, osseous, inflammatory, or miscellaneous.[2] Congenital lesions account for two-thirds of all presacral tumors.[1] Congenital lesions are more common in females and are usually benign; they include developmental cysts, sacrococcygeal chordomas, tailgut cysts, anterior meningoceles, and teratomas.[1]

Usually, tailgut cysts are asymptomatic and are incidentally discovered. However, symptomatic patients may present with a variety of symptoms, including rectal fullness, constipation, lower abdominal pain, or unexplained fecal/urinary incontinence. The initial presentation could be delayed until complications develop, including infection and abscess formation with fistulization, bleeding and malignant degeneration.[3] Many studies have shown that surgical excision is the definitive management method for these cysts.[4,5] However, nonsurgical management is still a controversial option.

We report here a case of an adult female who was diagnosed with recurrent tailgut cyst and underwent multiple surgical and nonsurgical procedures over a period of 17 years.

2 | CASE REPORT

This is a case of a 55-year-old female patient with no pre-existing medical illnesses, whose first presentation to the clinic was in 2001 with anal pain and mass. On examination, a retrorectal fullness was felt posteriorly with no signs of inflammation or fistulization. The anal sphincter tone was normal with an empty rectal vault. The mass was suspected to be an abscess and was aspirated under computed tomography (CT) guidance and surgical excision was planned, using a posterior approach. Drained fluid was thick and resembled pus. A biopsy of the wall of the abscess revealed the presence of multiple fragments of tissue lined by columnar epithelia with goblet cells and focal cilia formation. Bundles of haphazard smooth muscles were also noted, and the underlying stroma was focally edematous and infiltrated by mononuclear cells. These findings indicated a tailgut cyst.

A year later, the patient presented again with anal pain. Given her history and pathology, a CT scan was performed. A pelvic CT scan showed multiloculated cystic lesions posterior to the anus and rectum, slightly displacing the rectum anteriorly and indenting its posterior wall.

The patient underwent excision by a posterior approach to avoid injury of the rectum and anal sphincter. We found multinodulated cysts branching laterally and ascending 10 cm in the retrorectal space, while the anterior wall was attached to the wall of the anus and the rectum, forming an endless tree root-like structure. A small number of cysts attached to the posterior wall of the rectum were left untreated, due to the high risk of injury.

In 2006, the patient underwent excision of two main presacral cysts through the same approach. A thick yellowish odorless material was found. De-roofing of the remaining cysts was performed, which were also attached to the posterior rectal wall.

In 2007, a follow-up endo-rectal ultrasound report showed a multiloculated cystic structure of approximately 4.7 × 4 cm with internal echo, posterior to the rectum. No definite adjacent enlarged lymph nodes and no other pelvic fluid collection were found while the patient was asymptomatic.

In April 2008, the patient presented for the fourth time with the same complaints. A CT scan of the pelvis showed a large (7.6 × 4.6 × 5.5 cm) right perianal low-attenuation area with no evidence of fistula communication to the anal canal. This time, the patient opted for a less invasive approach. She underwent a CT-guided cyst drainage with an aspiration of about 15 cc of yellowish/reddish fluid, followed by injection of alcohol (10 cc). Biopsy revealed benign squamous tissue in a background of neutrophils, histiocytes, and degenerated cyst content. No malignant cells were seen.

The patient was symptom-free for almost 10 years when she came back to our clinic with dull nonradiating anal pain associated with a posterior anal mass. A pelvic MRI demonstrated a well-defined encapsulated multicystic mass measuring 7 × 6 × 6 cm. There was no evidence of invasion of the rectum nor lymph node involvement. Again, the patient preferred the less invasive method of treatment and chose to undergo another CT-guided drainage. No alcohol was injected since the aspirated fluid was thick and highly suspicious of pus. Cultures were negative for bacteria and acid-fast bacilli. Histological examination showed neutrophils, histiocytes, a few lipophages, and some calcified structures in a background of necrotic debris with no malignant cells. The patient was discharged on broad-spectrum antibiotics the next day. The patient was seen after 1 week, when she was found to be well, with no complaints or symptoms. A follow-up MRI after 1 month was planned. See Figures 1 and 2.

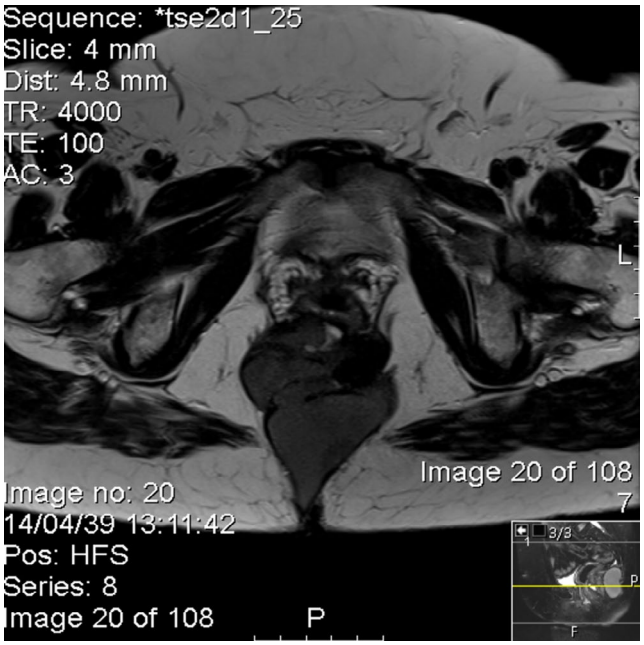

FIGURE 1 Pelvic MRI showing multicystic mass in the presacral space (sagital view)

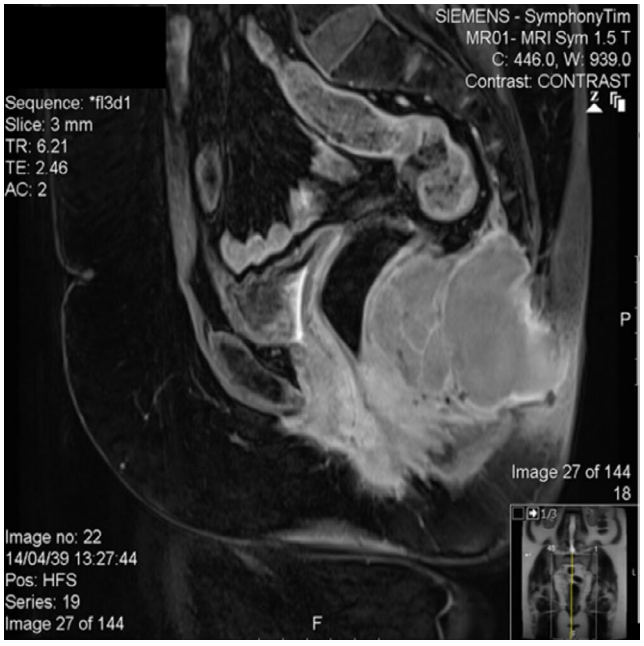

FIGURE 2 Pelvic MRI showing multicystic mass in the presacral space (axial view)

3 | DISCUSSION

Tailgut lesion is a rare type of congenital cyst. During the early weeks of gestation, the embryo has a true tail that lies caudal to the site of the subsequent formation of the anus. The primitive hindgut extends into this tail, hence the name

"tailgut."[5,6] Normally by the eighth week of gestation, the tailgut and the tail become atrophied.[3] Sometimes, the tailgut remnants persist, giving rise to tailgut cyst, also termed retrorectal cystic hamartoma.[5,7]

Tailgut cysts should be differentiated from other presacral cysts and masses, including epidermoid cyst, dermoid cyst, teratomas, rectal duplication cyst, anal gland cyst, cystic meningocele, and lymphangioma.[5,8] Also, infected tailgut cysts should be differentiated from pilonidal cyst, perianal fistulas, and abscesses.[9] This differentiation is very important because an abscess must be treated by a combination of drainage and antimicrobials, while tailgut cysts should be surgically excised.[10] A retrorectal cyst should be suspected in patients with recurrent pelvic abscesses and repeated operations for anal fistula without any underlying obvious pathology.[11]

In this case report, the patient was a middle-aged woman, which is consistent with previous studies that state a predilection for the adult age (despite being congenital) and a male to female ratio of 1:3.[5,9,10,12-14] The cause for this gender difference is unknown but may be explained by incidental discovery during vaginal examination for pregnancy and childbirth. It has also been reported in other age groups, including infancy.[5,12]

Approximately half of all tailgut cysts are asymptomatic and found incidentally.[15] Manifestations arise when the cyst exerts local pressure on surrounding structures, causing constipation and urinary disorders.[5,9,16-21] Also, symptoms arise when complications (in the form of bleeding, infection or malignant degeneration) ensue.[16,17,19,20] Infection is the most common complication (40%-50%) and presents with pelvic pain, and sometimes with local abscess and perianal fistulas.[16,17,20,21] Malignant degeneration has been reported in about 7% of tailgut cysts.[12,22] Our patient was symptomatic, probably due to the presence of multiple cysts and their large size. Nonspecific symptoms, including pain, are the most common complaints of tailgut cyst patients.[5]

The length of tailgut cysts typically range from 2 to 12 cm.[21,23] The cyst in the present case was relatively large. Similar sizes have been reported in previous case studies.[10,24]

A careful digital rectal examination is the initial examination of choice. In a series of 120 cases from Mayo clinic, 97% of the masses were palpable,[5,25,26] as in the present case.

Transrectal ultrasound may be useful in demonstrating the integrity of the layers of the rectum to show absence of invasion in a benign lesion.[27] However, cross-sectional imaging techniques are more reliable at defining the location of the cyst(s) in the retrorectal space.[28] MRI is a useful technique to evaluate pelvic disorders because of its multiplanar imaging capability and its accurate soft tissue contrast.[13,29]

Tailgut cysts may recur despite surgical treatment,[5] as in our patient who suffered multiple episodes of anal pain. Recurrence may be explained by the presence of multiple small cysts that are left after excision of the main cyst.

The relative rarity of these lesions, along with the nonspecific symptoms, can lead to misdiagnosis of these cases.[30,31] Such a scenario was reported in the case study conducted by Killingsworth and Gadacz,[11] as the patient was initially misdiagnosed with a perirectal abscess, which was treated by transrectal incision and drainage.

There is almost a consensus that surgical excision of resectable lesions followed by a complete histological examination is the recommended procedure for the treatment of tailgut cysts. However, unresectable lesions will usually require biopsy with a possible risk of seeding of malignant cells or infection of the previously sterile cysts.[4,21,32] Also, fine-needle aspiration under endosonographic guidance is not risk-free.[33]

Malignant transformation has been reported in a few cases,[34-37] including carcinoid tumor and adenocarcinoma. Careful histological examination should be conducted on the extracted surgical specimen. Fortunately, in our patient, histopathologic examination ruled out the possibility of malignant changes.

Tailgut cysts should be surgically excised,[31] even when discovered incidentally in asymptomatic patients, as silent masses can potentially present later with infection, recurrent perianal fistulas, or malignant transformation.[4,9] Surgical resection serves to provide a convenient specimen for histopathological examination, relieve symptoms caused by local compression, avoid subsequent infection, and rule out malignant transformation.[38]

Multiple surgical approaches have been described in the literature, including anterior (transabdominal), posterior, and combined pelvic approaches.[30] Posterior approaches, including trans-sacral, intersphincteric, and parasacrococcygeal are recommended for low-laying tumors below the level of S4.[39] Literature noted that an intersphincteric-approach excision preserves sphincter function, whereas a parasacrococcygeal approach is preferred in lesions with a suspicion of malignancy.[40] Furthermore, minimal invasive surgery (laparoscopic) has been reported as an effective approach in the narrow pelvis with low-laying retrorectal lesions.[40] Surgical decision depends on the degree of involvement of the adjacent structures, as invasion would mandate en bloc resection to prevent recurrence and possibility of malignant transformation.[5,15] Although surgical excision is the best management option, other nonsurgical approaches, including incision and drainage (I&D), with or without alcohol injection, might be adopted for high-risk patients and patients who prefer less invasive solutions as is the case in our case report. A case reported in 2005 of a patient with retrorectal cystic hamartoma showed a failure of I&D to prevent recurrence within months; however,

complete resolution was attained by using complete surgical excision.[11]

We reported this rare case to increase awareness in the medical community about this rare condition and highlight the different treatment approaches.

4 | CONCLUSION

Alcohol injection, although not curative nor replacing the classical surgical excision, can be considered as an alternative method of treatment for tailgut cysts for special cases who are unwilling or unable to withstand a surgical intervention.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

FA, BA and RA: involved in literature review, writing the introduction, case, and discussion. NM and ZA: involved in supervision and editing on the introduction, case, and discussion. ZA is the principal investigator.

ORCID

Farrah Aljuhani https://orcid.org/0000-0002-0556-4070
Nadim Malibary https://orcid.org/0000-0002-1405-331X

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